Systematic Review

HAMMILL INSTITUTE ON DISABILITIES

Communication Disorders Quarterly I–8

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DOI: 10.1177/1525740119870772

cdq.sagepub.com

Description of Multilingual Participants Who Stutter: An Update 2011-2018

Danielle R. Werle, MA, CCC-SLP¹, Courtney Byrd, PhD, CCC-SLP¹, and Geoffrey Coalson, PhD, CCC-SLP²

Abstract

Coalson, Peña, and Byrd published a systematic review that demonstrated significant limitations in the language profile descriptions for multilingual participants who stutter for studies that were completed through September 2011. Given the average number of investigations of multilingual speakers who stutter has shifted from one study every 5 years to three and a half studies every year, the purpose of this study was to determine if these limitations in language profile descriptions have persisted. A systematic search of the stuttering literature between October 2011 and October 2018 was conducted to identify the number of published studies focusing on multilingual participants. Descriptors of participants' language history, function, and proficiency were assessed within and across studies. The quality of these descriptions remains insufficient. Despite the marked increase in published research, these persisting limitations in language profile descriptions continue to compromise our understanding of the manifestation of stuttering in more than one language.

Keywords

stuttering, English as a second language (ESL)/bilingualism/dialects, English/languages, research

Introduction

Stuttering is a complex speech disorder that has been identified in monolingual speakers and speakers of more than one language worldwide. However, the number of peer-reviewed studies conducted within this clinical population, particularly with regard to multilingual speakers, is limited. In a systematic review of the literature, Coalson, Peña, and Byrd (2013) reported that from January 1900 to September 2011 there were only 23 peer-reviewed studies published that examined the prevalence, treatment, and/or characteristics of stuttering in multilingual speakers. In addition, the number of participants within each of these studies was restricted to either single subject or a number that was too low to generalize results. Given the challenges in recruiting a sufficient number of multilinguals who stutter with similar language profiles within a single study, it is unlikely that our understanding of this unique clinical population will advance without meta-analysis (see Ntourou, Conture, & Lipsey, 2011) or data aggregation (see Brundage, Corcoran, Wu, & Sturgill, 2016). To facilitate such cross-study comparisons, adequate and comparable descriptions of participants' language profiles are needed.

Multilingualism and Stuttering: 1900-2011

For the better part of the 20th century (1900-2011), there has been a paucity in the research related to the prevalence, treatment, and characteristics of developmental stuttering in

speakers of more than one language. In terms of prevalence, a select number of studies identify multilingualism as a potential risk factor for the development of stuttering in children (e.g., Howell, Davis, & Williams, 2009; Karniol, 1992; Mohamadi, Nilipour, & Yadegari, 2008; Stern, 1948). However, these claims have been disputed due to methodological concerns (e.g., Byrd, Bedore, & Ramos, 2015; Byrd, Watson, Bedore, & Mullis, 2015; Packman, Onslow, Reilly, Attanasio, & Shenker, 2009).

Research describing the characteristics of stuttering in multilinguals has been largely operationalized via "language dominance" wherein multilingual participants are classified into discrete categories based on their strength in each language. Across studies, data do not demonstrate a consistent relationship between stuttering and language dominance. Earlier studies suggest that balanced bilinguals stutter more in one language, suggesting language-specific differences in stuttering frequency (Bernstein Ratner & Benitez, 1985; Dale, 1977; Nwokah, 1988). Four subsequent studies, however, provide counterevidence indicating

Corresponding Author:

Danielle R. Werle, The University of Texas at Austin, 2504A Whitis Avenue, #A0720, Austin, TX 78712, USA. Email: danielle.werle@utexas.edu

¹The University of Texas at Austin, USA ²Louisiana State University, Baton Rouge, USA

more stuttering in the speakers' less-dominant language (Ardila, Ramos, & Barrocas, 2011; Jankelowitz & Bortz, 1996; Lim, Lincoln, Chan, & Onslow, 2008; Roberts, 2002), and one study indicated greater stuttering in the dominant language (Carias & Ingram, 2006).

Conflicting outcomes within the multilingual stuttering literature may reflect, at least in part, the divergent manner of language profile descriptions across studies. Coalson et al. (2013) reviewed the breadth and depth of descriptions of multilingual participants across the stuttering literature. Studies were examined with respect to Grosjean's (2004) recommended framework for describing multilingual participants, which included three primary factors: language history, language function, and language proficiency. Language history is defined as the age and conditions in which the speaker was exposed to their second/non-native language (L2, first/native language: L1). Language function is defined as the amount or frequency that each language is currently used across specific settings and interlocutors. Language proficiency is defined as the speaker's overall ability to produce and understand each language in verbal or written form. In terms of breadth of description, the most frequently reported factor across 23 studies was language proficiency, followed by history, and function. However, nearly one-half of the 23 studies (n = 10, 43%) included minimal information regarding any of these factors thereby limiting understanding and cross-study comparison of the bilingualism of the participant(s). In addition to infrequent reporting of each factor within and across studies, the information used to determine each of these factors, if included, varied considerably across studies (i.e., 13-29 different criteria per factor) and were largely subjective in nature.

According to Grosjean (2004), the heterogeneity of language history, function, and proficiency between and within multilingual speakers is a significant methodological concern when attempting to interpret finding across, or even within, studies. Linguistic ability and use vary based on the conditions of initial exposure, frequency of use/ demand, and underlying speech and language capacities. From a psycholinguistic perspective, each factor has been found to uniquely influence specific speech and language skills in multilingual adults (articulation: Fowler, Sramko, Ostry, Rowland, & Hallé, 2008; phonology: Flege, Yeni-Komshian, & Liu, 1999; semantics: Newman, Tremblay, Nichols, Neville, & Ullman, 2012; and morphosyntactic: Birdsong & Molis, 2001) and particularly children (e.g., Bedore et al., 2012; Bohman, Bedore, Peña, Mendez-Perez, & Gillam, 2010; Hoff et al., 2012; Thordardottir, 2017). Language use may be modified based on communicative context, task, or emotional content of message (e.g., code-switching: Paradis & Nicoladis, 2007; language preference: Optiz & Degner, 2012). Thus, to account for the differences in language profiles, participant descriptions cannot be limited to one factor, rather they should

include detailed descriptions of history, function, and proficiency.

Further complicating matter is the co-occurring variability of stuttering itself. A large body of data support that, similar to multilingualism, stuttered speech will manifest differently based on the speakers' underlying speech and language abilities, particularly in children (e.g., Bloodstein, 2006; Ntourou et al., 2011). An equally large body of research indicates that stuttering will differ across situations, listeners, and content of the intended message (Bloodstein & Bernstein Ratner, 2008). These overlapping aspects of multilingualism and stuttering make it difficult to attribute any observable difference in speech patterns to language status (mono- vs. multilingual) or talker status (person who stutters vs. typically fluent). Furthermore, reliance on any single criterion of a participant's language profile is likely too narrow to confidently attribute outcomes to language status. A narrow understanding of multilingualism in the field of fluency puts children at risk for false positive diagnosis of stuttering (Byrd, Haque, & Johnson, 2016; Byrd et al., 2015). Given recent research has documented the wide variety of disfluent speech in children who do not stutter and who speak multiple languages (Byrd et al., 2015), it is critical to comprehensively describe language profiles to determine if the presence of disfluent speech reported in these studies is secondary to language history, function, or proficiency, or if a fluency disorder is in fact present. The only way to conclusively make these decisions and judgments is with comprehensive description of language profiles.

Rationale for This Study

Several factors motivate the need for the present replication of Coalson et al. First, the number of people who speak more than one language in the United States in 2015 exceeded what was projected in 2010, up by approximately 3 million people, and is likely to exceed the estimated number multilinguals projected for 2020 (approximately 64-68 million people; Ortman & Shin, 2011). This rapid growth has sparked a notable increase in the discussions and investigations of bilingualism. Review of conference programs and proceedings from three fluency-related research forums from 2011 to 2017 (American Speech-Language-Hearing Association Convention [ASHA], 2011-2017; International Fluency Association [IFA], 2012, 2015; Oxford Dysfluency Conference [ODC], 2014, 2017) list not less than 44 oral and poster presentations that address multilingualism and stuttering (ASHA, n = 27; IFA, n = 12; ODC, n = 8). Upon cursory review of the recent literature, the need for this study was apparent, as there has been a marked increase in peer-reviewed publications with the average shifting from one study every 5 years related to stuttering in bilinguals to three and a half studies every year. Although this is an encouraging trend, review of the peer-reviewed published literature is necessary to Werle et al. 3

determine whether or not along with this marked increase there has been an increase in the participant descriptions that would facilitate understanding of the manifestation of stuttering in bilinguals as well as reliable and valid cross-study comparisons. One would anticipate that such a significant increase in quantity of empirical investigations into bilingualism would be accompanied with an equivalent increase in the quality and thoroughness of participant descriptions. Furthermore, with the recent data to suggest that bilingual children who are typically fluent may be at risk for false positive identification as stuttering (Byrd, Bedore, et al., 2015, Byrd, Watson, et al., 2015; Byrd et al., 2016), there is a critical need to examine whether the participants included in the previous studies are described in a manner that adequately conveys their bilingualism and how their acquisition of that second language is of relevance to their speech disfluency and/or stuttering. Thus, the purpose of this study was to determine if these limitations in language profile descriptions with regard to language history, function, and proficiency have persisted, despite the significant increase interest and empirical explorations. If, as previously reported, there continues to be a lack in breadth and depth of participant description, then similar to the call to require journals to include effect sizes (see Wilkinson and the APA Task Force on Statistical Inference, 1999), this study will serve as a call to require key descriptors of participants' language profiles in future studies to improve our understanding of stuttering in bilinguals.

Method

A systematic search was conducted to identify the number of studies using multilingual participants who stutter between September 2011 and October 2018. Descriptions provided for multilingual participants who stutter in each identified study were examined to assess the quality of description relative to three primary factors: language history, language function, and language proficiency.

Search Procedure and Terms

Two online databases were searched to identify qualifying studies: (a) EBSCO, including PsycINFO, Academia Search Complete, Book Review Digest Plus, Communication and Mass Media Complete, eBook Collection, Education Source, ERIC, Fuente Academica Premiere, Healthsource: Consumer Edition and Nursing/Academic Edition, History of Science, Technology, and Medicine, Humanities full text, Library and Information Science Source, MEDLINE, Sciences Psychology and Behavioral Collection, PsycARTICLES, and Science and Technology Collection as well as (b) Google Scholar. Search terms included combinations of the following terms: bilingual, bilingualism, multilingual, multilingualism stuttering, stutter, stammer, and stammering. This yielded a total of 16 unique search term combinations. Both databases included search terms in the entire text. The search was filtered for publications available from October 2011 to October 2018, to extend beyond the search completed by Coalson et al. (2013).

Inclusionary Criteria and Review Procedure

The 16 search terms across two bases yielded 4,281 unique items. The titles and abstracts of these items were reviewed by the first author and a trained research assistant for relevance according to the inclusionary and exclusionary criteria. Studies were considered eligible for review if the following criteria were met: (a) included multilingual participants who stutter, (b) published in 2011 or later, (c) provided original data, and (d) full text was published in English or translated into English. Articles were excluded if (a) they were unpublished manuscripts, theses, conference proceedings, or posters, (b) participants who stutter did not speak two or more languages, (c) the articles were reviews of the literature, or lacked original data, and (d) full text was not available in English. Following review of titles and abstracts for adherence to inclusionary and exclusionary criteria, 92 of the original 4,281 items were included for full detailed review of methods.

Thirty one of these 92 were unpublished theses or conference proceedings. Twenty four contained multilingual participants who did not stutter, four papers were either not published in or not translated to English. Finally, eight papers lacked original data. This resulted in 25 studies being included in the final review of participant characteristics. For the final analysis, a full review of the methods, results, and discussion assessed all participant characteristics, as well as the purpose of each study. All 25 unique studies identified during the review are denoted with an asterisk (*) in the references.

Results

Since September 2011, 25 additional studies have been published using multilingual participants who stutter, or approximately three and a half studies every year. These findings indicate a substantial increase in published data on this topic relative to Coalson et al. (2013), who reported 23 studies between 1900 and 2011, or approximately one study every 5 years. Nineteen of the 25 studies (76%) were considered descriptive—higher than the number of descriptive studies reported in Coalson et al. (2013; 14 of 23 studies, or 61%). Six studies focused on treatment of stuttering in bilingual participants (24%), up from 22% in 2013, and no studies focused on the prevalence of stuttering in the multilingual population, compared with the four studies reported in Coalson et al. (2013; 17%).

Breadth of Language Profiles

Examination of breadth included comparison of (a) how often each factor was included across studies and (b) the

Language Profile	1900-2011 ^a (n = 23)	%	2011-2018 (n = 25)	%	Trend
Across studies					
History	13	56	16	64	Increase
Function	10	43	13	52	Increase
Proficiency	16	70	14	56	Decrease
Within studies					
All three factors	7	30	11	44	Increase
Two of three factors	6	26	3	12	Decrease
One of three factors	6	26	4	16	Decrease
Zero of three factors	4	17	7	28	Increase

Table 1. Breadth of Participant Description in Stuttering Literature Before 2011 and From 2011 to 2018.

number of factors included within each study. Table 1 provides a detailed summary of each factor within and across each of the 25 studies compared with data reported by Coalson et al. (2013).

Across studies. Of the 25 studies, language history was reported most frequently, followed by proficiency and function. History and function were reported with higher frequency compared with the 2013 study (Coalson et al., 2013), whereas proficiency was reported with lower frequency. Overall, more than half of the 25 studies provided some information about either the participants' language history, function, and proficiency (64%, 52%, and 56%, respectively), indicating a slight overall increase from the frequencies with which these factors were reported in the 2013 study (56%, 43%, and 70%, respectively).

Within studies. Of the 25 studies, nearly half provided information regarding all three factors (44%), up from one-third reported in 2013 (30%). However, the proportion of studies that provided no information was larger (28%; 6 of 25 studies) than the 2013 study (17%; 4 of 23 studies). The number of studies that provided information for one or fewer factors in this study (44%; 11 of 25 studies) remained relatively similar to the 2013 study (43%; 10 of 23 studies). These 11 studies also account for almost half of the total number of multilingual participants who stutter (n = 168 of 362, 46%).

Depth of Language Profiles

Number of different descriptors. As illustrated in Table 2, and similar to Coalson et al. (2013), the three primary factors were determined using a wide range of information. Language history was reported with the greatest number of descriptors (15 different descriptors), a notable decrease compared with the 2013 study (29 different descriptors). Language function and language proficiency were reported using 11 different descriptors, roughly equivalent to the 2013 study (13 different descriptors). Further details on

specific descriptors used to determine language history, function, and proficiency are provided in Supplemental Appendices A to C, respectively.

Consistency of descriptors. Overall, the consistency of descriptors used across studies mirrored the results reported in Coalson et al. (2013, see Table 3), particularly language history and proficiency. Of the 16 studies that included information about language history, two descriptors were used most frequently: age/years of initial L2 exposure and language used at home. These descriptors were also most frequently reported in the 2013 study, with a notably higher proportion of studies (69%; 11 of 16) that included age/years since initial L2 exposure.

Although these data indicate a positive shift toward a common descriptor for language history, the remaining studies (5 of 16; 31%) relied on information other than time since initial L2 exposure, and five of the 16 studies (31%) relied solely on qualitative information (e.g., language used at home; language used at school). It should also be noted that nine of the 25 total studies (36%) provided no information regarding language history.

Of the 14 studies that included information about language proficiency, two qualitative descriptors were used most frequently: overall judgment of examiner and self-reported proficiency across modalities. These are similar to the two most frequent proficiency descriptors used in the 2013 study: judgment of the speaker or examiner and self-ranked spoken proficiency. Fewer studies reported overall examiner judgment (36%; 5 of 14) compared with the 2013 study (56%; 9 of 16), and a slightly greater percentage of studies reported selfranked proficiency (29%; 4 of 14) compared with the 2013 study (19%; 3 of 16). Although these data indicate a slight shift toward self-rated proficiency as a common descriptor for language proficiency, 10 of the 14 (71%) studies used criteria other than self-ranked abilities, and 11 of the total 25 (44%) studies provided no information regarding proficiency. Furthermore, all five studies which reported examiner judgment used this as their sole descriptor of proficiency, and two of these five studies failed to rank L1 proficiency.

^aData obtained from most recent systematic review conducted by Coalson, Peña, and Byrd (2013).

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Table 2. Depth of Multilingual Participant Description in Stuttering Literature Before 2011 and Within the Last 7 Years.

Primary Factor	1900-2011 ^a				2011-2018				
	Number of descriptors	Consistency of descriptors				Consistency of descriptors			
		Most frequent criteria	Туре	Number of studies	Number of descriptors	Most frequent criteria	Туре	Number of studies	
History	29	Overall home lang	Qual	9/13		Age/yrs L2 exposure	Quant	11/16	
		Age/years L2 exposure	Quant	6/13		Overall home lang	Qual	6/16	
Function	13	Estimation of examiner	Qual	7/10	12	Preferred home lang	Qual	4/13	
		Preferred lang work/ school	Qual	6/10		% LI and L2 at home/ school	Quant	3/13	
Proficiency	13	Overall judgment of speaker/examiner	Qual	9/16	Ш	Overall judgment of examiner	Qual	5/14	
		Self-ranked proficiency (speaking)	Qual	3/16		Self-reported proficiency (all modalities)	Qual	4/14	

^aData obtained from most recent systematic review conducted by Coalson, Peña, and Byrd (2013); review terminated September 2011. Qual: criteria determined via qualitative or subjective measure; Quant: criteria determined via quantitative or objective measure.

Language function demonstrated the most notable departure from the previous review. Of the 13 studies that included information regarding language function, preferred language at home and percentage of L1 and L2 used at work/school were the most frequently reported. This pattern differs from the 2013 study, which found function most frequently described by using qualitative measures. That being said, language function was also the least consistently described of the three primary factors, with only four of 13 studies (31%) using a common descriptor. Twelve of the total 25 studies (48%) provided no information about language function.

Discussion

The present systematic search of the literature from October 2011 to October 2018 revealed a notable increase in the number of peer-reviewed publications that include participants who stutter who speak two or more languages. Over the past 7 years, there has been an average of approximately three and a half studies per year, compared with the average of one study every 5 years from January 1900 to September 2011. Despite this increase, the quality of participant description with respect to language history, function, and proficiency remains inconsistent.

Breadth and Depth of Descriptions

In terms of breadth, the number of studies providing some information for language function and history increased, as well as the percentage of studies including all three primary factors. This increase may reflect the number of studies that were descriptive in nature, or perhaps the number of studies that focused on children rather than adults. These reasons notwithstanding, a larger number of studies that report wider

range of descriptive factors for multilingual participants than the 2013 report should be considered an encouraging trend. That being said, almost half of the literature described the participants' language profile using only one of these factors or provided no description of multilingual language abilities. Unfortunately, these studies also account for almost half of the total number of multilingual participants who stutter (n = 168 of 362, 46%). The lack of adequate description of language abilities limits our ability to understand how stuttering manifests during the acquisition of multiple languages and how the basic characteristics of stuttered speech (e.g., function/content words: Gkalitsiou, Byrd, Bedore, & Taliancich-Klinger, 2017; Schäfer & Robb, 2012) may differ across languages due to differences in specific aspects of language profiles (Hoff et al., 2012).

In terms of depth, each factor was characterized by at least 11 different descriptors across studies, and no single descriptor was used in more than 44% of the studies. This is a slight increase from the 2013 review, but still reflects disagreement across studies when defining each factor. Language history was determined using the most varied criteria, including 15 different descriptors, but most frequently described via time since initial L2 exposure. Language function did not appear to follow any pattern related to the demographic of participants. As illustrated in Supplemental Appendix B, information was collected from a variety of settings (e.g., home, work, and religious events), but rarely were these settings reported more than once across studies. Consistent with studies reviewed in Coalson et al. (2013), language spoken at home as well as percentage of L1/L2 spoken at home or school remain the most frequent descriptors of language function. However, these descriptors were reported in less than 30% of the studies. Underreporting of language function is of concern, given the relative ease with which this information may be obtained and insight it

provides for language dominance (Bedore et al., 2012). Language proficiency was described using the most restricted range of descriptors. However, no single metric was used in more than half of the 14 studies that included information about proficiency.

It should be noted that the use of multiple criteria to describe proficiency, or any of the three primary factors, is not necessarily a criticism of the literature. For example, a subjective rating scale of proficiency that covers different linguistic domains, combined with a formal assessment of one or more specific domains of interest, may provide researchers with a relatively comprehensive assessment of linguistic abilities. In fact, self-rated proficiency has been found to correlate reasonably well with third-party ratings of proficiency (5-point Likert-type scale: Delgado, Guerrero, Goggin, & Ellis, 1999; Hasson, 2008) and may serve as a simple but effective metric to compare multilingual participants across studies. Nevertheless, even within this self-reported proficiency there was some discrepancy. For example, two studies only reported proficiency ratings for the participants' second language, rather than both known languages. Although these participants were considered sequential bilinguals, the assumption that L1 proficiency remains stable after L2 acquisition is unsubstantiated (Kohnert, Bates, & Hernandez, 1999; Linck, Kroll, & Sunderman, 2009) and further illustrates the need for comprehensive data, such as language history, and perhaps secondary data, such as language stability.

Future Research

Since the publication of Coalson et al. (2013), which sets forth recommendations for inclusion of language history, function, and proficiency in investigations involving multilingual participants who stutter, some improvements have been noted with respect to the amount of and consistency of language profile information in recent literature. From the review of the current literature base, trends have emerged with respect to common metrics that used to improve the consistency and comprehensive nature of language profiles. It is recommended that future research consider the use of the following characterizations as metrics for each primary factor. These metrics were chosen based on the quality with which they represent bilingual language experiences and abilities and also accessibility to researchers.

To quantify language history, in accordance with bilingual literature and current trends in the stuttering literature, use of length of time as well as contexts participants are exposed to each language should be recorded. To capture present language function, especially as it relates to language dominance, reports of percentage of L1 and L2 input and output across weekday and weekend activities should be collected from the participant and/or caregivers. Alternatively, published language function questionnaires are easily accessible and replicable tools for researchers

the Alberta Language and Development (e.g., Questionnaire [Paradis, Emmerzael, & Duncan, 2010]; the Bilingual Language Profile [Birdsong, Gertken, & Amengual, 2012], for a comprehensive list, see Table 1 in Coalson et al., 2013). To establish language proficiency, available published assessments, such as the Bilingual English Spanish Assessment (BESA; Peña, Gutierrez-Clellen, Iglesias, Goldstein, & Bedore, 2014), would be ideal. In the absence of a standardized measure translated to the appropriate language or combination of languages, a comprehensive self-report should be employed. A selfreport rating should encompass all languages spoken, as well as multiple modalities.

Acknowledgments

This project was complete with endowed support provided through the Michael and Tami Lang Stuttering Institute and the Dr. Jennifer and Emanuel Bodner Developmental Stuttering Laboratory awarded to the second author. This project also fulfills part of the research requirement for the first author's receipt of the Department of Education, Language Evaluation and Development in Educational Research doctoral training fellowship. We would like to thank master's student and bilingual Speech-Language Pathologist, Alejandro Granados Vargas, for his assistance with the initial literature search.

Declaration of Conflicting Interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.

Supplemental Material

Supplemental material for this article is available online.

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